

## CASE REPORT

## Multiple metastatic tumors in the oral cavity

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### ABSTRACT

Metastatic lesions to the oral region are uncommon and account for approximately 1% of all malignant oral tumors. In 25% of the cases, oral metastases are found to be the first sign of the metastatic spread; and in 23% of the cases, it is the first indication of an undiscovered malignancy at a distant site. Metastases to oral soft tissues are even less frequent than jaw bones. Because of its rarity, the clinical presentation of a metastatic lesion in the oral cavity can be deceiving, leading to a misdiagnosis of a benign process; therefore, in any case where the clinical presentation is unusual, especially in patients with a known malignant disease, a biopsy is mandatory. Here, we are presenting a rare case of multiple secondary tumors in the attached gingiva in an otherwise apparently healthy patient with no other symptoms of the primary tumor. It subsequently led to the diagnosis of Pancoast tumor (bronchoalveolar carcinoma) metastasizing simultaneously to multiple sites in the oral cavity and bilateral adrenal glands.

**Key words:** Bronchoalveolar carcinoma, metastatic tumor, pancoast tumor

### INTRODUCTION

Oral metastatic lesions from distant tumors are uncommon and mainly involve the bony structures, whereas metastases to soft tissues are extraordinarily rare. In most patients, the primary tumor is already known before the oral metastatic lesions appear.<sup>[1]</sup> This particular case is worth reporting, because of extremely rare nature of simultaneous metastasis of Pancoast tumor to the adrenal gland and gingiva and in spite of the wide spread metastasis the patient did not present with any other symptoms, except for the presence of recurrent gingival lesions. The clinical appearance in this case was of a gingival mass that could have been misdiagnosed as a hyperplastic lesion associated with a tooth or as a reactive process. In many cases diagnosis can be difficult and requires patient's remote medical history, biopsy for histological diagnosis as well as some target organs' investigations, such as brain, lungs, liver, to establish the correct diagnosis.

### CASE REPORT

A 49-year-old male patient reported to the department of

Oral and Maxillofacial Surgery, Government Dental College, with a painless sessile growth of size 2 × 3 cm on the lingual attached gingiva in relation to mandibular incisors of one month duration. Clinically, only four mandibular anterior teeth were present which includes left incisors, right central incisor, and canine. Rest of the teeth were extracted due to chronic generalized periodontitis. Clinical examination revealed the lesion to be soft to firm in consistency with no bleeding on probing. Grade 3 mobility of anterior teeth was found except for the left lateral incisor which was grade I mobile. The overlying mucosa appeared blanched and no evidence of ulceration was noticed. Oral hygiene was poor.

Radiographic examination showed extensive bone resorption in relation to the mandibular anterior (left lateral incisor to right lateral incisor) region with a floating tooth appearance [Figure 1]. A tentative diagnosis of pyogenic granuloma was suggested by the oral surgeons as there were no other symptoms. All the teeth with grade 3 mobility were extracted and the lesion was excised.

After 20 days of initial excision, the patient came back with the presentation of recurrent lesion at the same site, i.e. on the gingiva in mandibular anterior teeth region in the extracted area [Figure 2] along with two other similar looking lesions on palate close to the alveolus on the left side in the molar region and in the maxillary tuberosity area on the same side [Figure 3]. On examination, the lesions were reddish in color, firm in consistency and bleeding on probing was also noticed. Submandibular and sublingual lymph nodes were enlarged and

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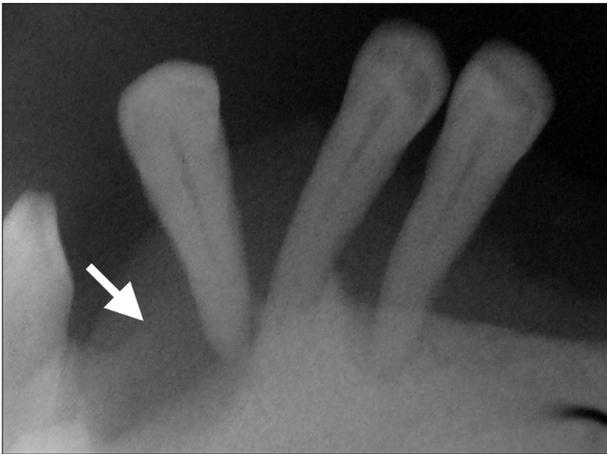


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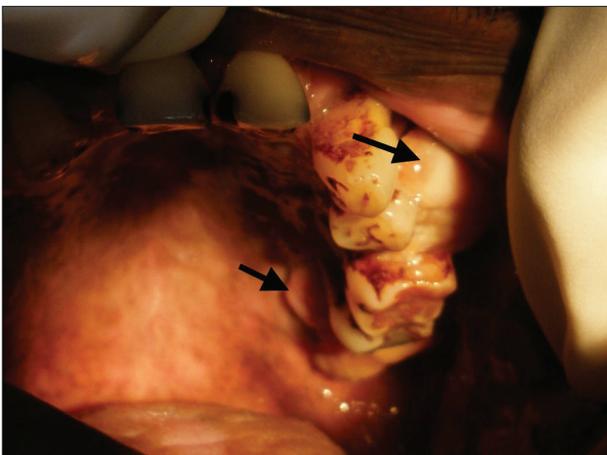
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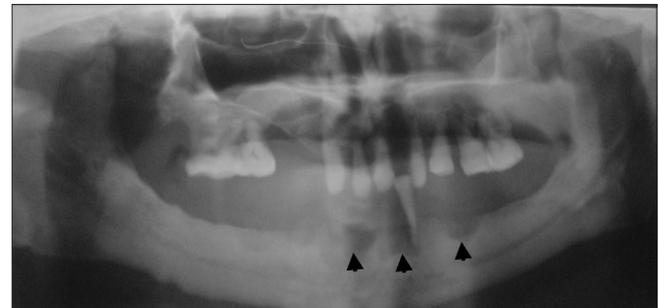
**Figure 1:** Extensive bone resorption in relation to the mandibular anterior teeth region with a floating tooth appearance



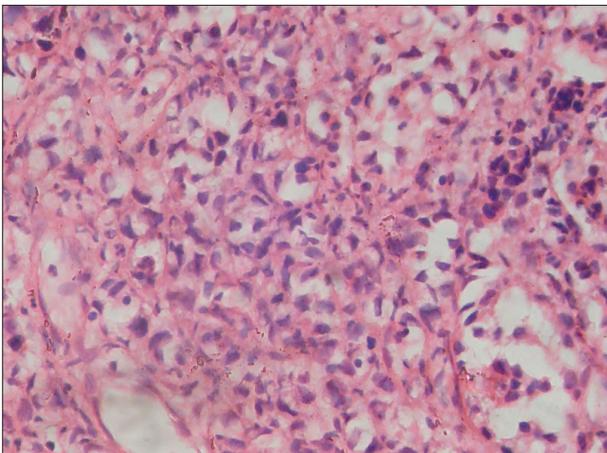
**Figure 2:** Recurrent lesion on the gingiva in mandibular anterior teeth region after the extraction



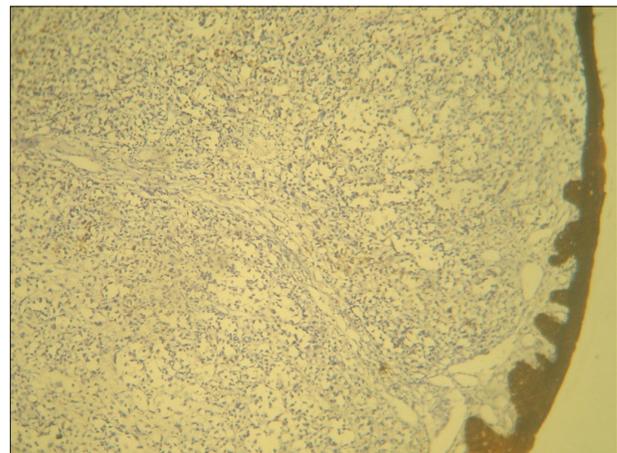
**Figure 3:** Lesion on palate close to the alveolus on the left side in the molar region and in the maxillary tuberosity area on the same side



**Figure 4:** Orthopantomogram revealing bone loss in the edentulous alveolus in relation to lower left and right incisor area and left canine – premolar region



**Figure 5:** H and E photomicrograph of magnification 40x showing large atypical hyperchromatic cells and some clear cells with nucleus pushed to one side



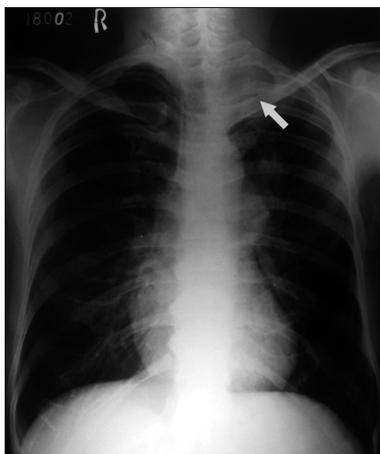
**Figure 6:** Immunohistochemistry for cytokeratin showing focal positivity, 4x

fixed, but no involvement of the supraclavicular, superficial and deep cervical lymph nodes. Patient was also complaining of weight loss and dull radiating pain in the left shoulder. Medical history revealed nothing.

On routine investigation, hemoglobin was seen to be reduced by two units over a period of 20 days.

Orthopantomogram revealed bone loss in the edentulous alveolus in relation to lower left and right incisor area and left canine – premolar region [Figure 4]

All the three lesions were excised in the department of oral surgery and sent for histopathological examination. On gross



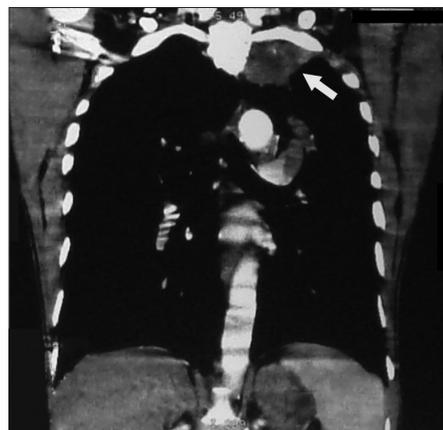
**Figure 7:** X-ray chest suggesting a homogenous opacity in the apex of left lung

examination, there were three bits of firm consistency; light brown in color. Size of the larger bit was  $1.5 \times 1 \times 1$  cm, and two smaller bits of size each  $1.5 \times 0.5 \times 0.5$  cm.

Histopathological examination of all the received specimens was done. Two out of three bits showed stratified squamous epithelium overlying connective tissue, which appeared torn at places which was thought to be a processing error. Underlying connective tissue showed some atypical cells with granular cytoplasm which were arranged in alveolar pattern. Large atypical hyperchromatic cells and some clear cells with nucleus pushed to one side were also seen. None of the cells had any resemblance to the native cells of oral mucosa [Figure 5]. The other two bits showed features of granulation tissue. The histopathological report was suggestive of metastatic malignancy. but we could not pinpoint the origin of the cell.

Patient underwent human immunodeficiency virus (HIV) test to rule out any HIV associated tumor, but it came negative. Other differential diagnoses were malignant minor salivary gland neoplasm, angioproliferative lesions, melanoma, and lymphoma. To rule out malignant salivary gland neoplasm, immunohistochemistry using P63 was done which again came negative. Immunohistochemical (IHC) markers, leucocyte common antigen (LCA) and CD 34, were used to rule out lymphoma and angioproliferative lesions, respectively. To rule out metastasis from lung and thyroid, thyroid transcription factor (TTF-1) was used which also came negative. Immunohistochemistry for cytokeratin showed focal positivity [Figure 6] which suggested metastasis from kidney or gastrointestinal tract (GIT) or lung.

Ultrasound of chest and abdomen showed hyperechoic bilateral adrenal mass. X-ray chest suggested a homogenous opacity in the apex of left lung [Figure 7] with destruction of underlying ribs. Right lung appeared clear. X-ray was suggestive of Pancoast tumor. Computerized tomography (CT) scan of chest and abdomen with IV contrast revealed an irregularly



**Figure 8:** The CT scan of chest revealing an irregularly and poorly enhancing soft tissue density mass in the apico-posterior segment of left upper lobe

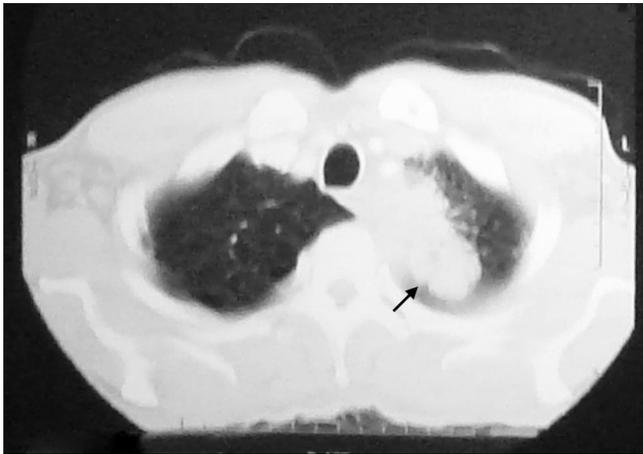
and poorly enhancing soft tissue density mass in the apico-posterior segment of left upper lobe of size  $6.2 \times 5$  cm [Figure 8]. Involvement of the pleura and left second rib posteriorly was noticed. Left apical soft tissue mass lesion measuring approx  $7 \times 5$  cm with destruction of the adjoining posterior aspect of left rib was also noted. Surrounding pneumonitis was present. The picture was suggestive of a primary in the lung. Well defined poorly enhancing areas were noted in the aorto pulmonary window and left hilum suggestive of enlarged lymphnodes of size  $3 \times 2$  cm. A left suprarenal mass was noted of size  $5 \times 3.2$  cm [Figure 9]. Right adrenal gland was also enlarged. There was no pleural effusion. Thyroid, trachea, bronchial tree, heart, and vascular structures appeared normal. Liver, biliary radicles, and spleen were appearing normal. There was destruction of sternum, mid dorsal vertebral body, and pedicle.

CT scan of head and neck region revealed destruction of left alveolar process with enhancing soft tissue component. There was destruction of floor of the left maxillary sinus with soft tissue component into it [Figure 10]. Infiltration into anterior aspect of left pterygoid and masseter muscles was noted. Multiple enlarged bilateral level IA, IB, and II nodes were noticed. A cyst measuring  $1.3 \times 1$  cm was noted within median glossoepiglottic fold.

The lesion was diagnosed as bronchoalveolar carcinoma of the left lung metastasized to bilateral adrenal glands and multiple sites in the oral cavity. Patient was referred to the Regional Cancer Centre, Trivandrum, for further treatment. Though chemotherapy was instituted, the patient died after nine months of initial diagnosis.

## DISCUSSION

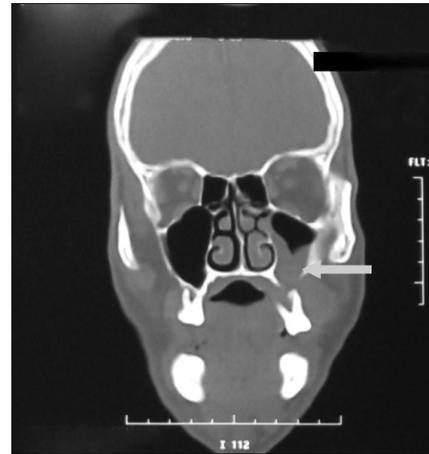
Metastasis to the oral cavity is very rare and represents only 1% of all neoplasm in the oral cavity. In 25% of the cases, oral metastases are found to be the first sign of the metastatic



**Figure 9:** The CT scan of abdomen revealing a left suprarenal mass

spread; and in 23% of the cases, it is the first indication of an undiscovered malignancy at a distant site. Oral soft tissues were less frequently affected than the jaw bones (1:2.5). In the oral soft tissues, the attached gingiva was the most commonly affected site (54%). The major primary sites presenting oral metastases were the lung, kidney, liver, and prostate for men; breast, female genital organs (FGO), kidney, and colo-rectum for women. In men, the lung was the most common primary site affecting both the jawbones and oral mucosa (22% and 31.3%, respectively) followed by the prostate gland in the jawbones (11%), and kidney in the oral soft tissues (14%). In women, the breast was the most common primary tumor affecting the jawbones and soft tissues (41% and 24.3%, respectively), followed by the adrenal and FGO to the jawbones (7.7%), and FGO to the soft tissues (14.8%).<sup>[2]</sup> Lung carcinoma normally metastasizes to the gingiva and kidney primary metastasizes to the jaw. According to Van der Waal, in most patients, the primary tumor was already known before the oral metastatic lesion appeared.<sup>[1]</sup> This particular case is worth reporting, because, in spite of the wide spread metastasis, the patient was unaware of the symptoms, except for the presence of recurrent oral lesions. Thus, a retrograde diagnosis had to be made by an experienced pathologist with the aid of all supporting investigations.

Pancoast tumor is a tumor in the apico-posterior part of the lung. It represents fewer than 5% of all primary lung cancers. Pancoast tumor can invade the brachial plexus, pleura, or ribs, causing shoulder and upper extremity pain and weakness or atrophy of the ipsilateral hand and Horner's syndrome (ptosis, miosis, enophthalmos, and anhidrosis).<sup>[3]</sup> More than 95% of Pancoast tumors are non-small cell carcinomas, most commonly squamous cell carcinomas (52%), or adenocarcinomas (23%), and large cell carcinomas (approximately 23%).<sup>[4]</sup> Bronchoalveolar carcinoma, a subtype of adenocarcinoma, is a malignant neoplasm of the lung arising from the epithelium of the bronchus or bronchiole in which patient presents with a variety of clinical manifestations like cough, weight loss, chest pain,



**Figure 10:** The CT scan of head and neck region revealed destruction of left alveolar process and floor of the left maxillary sinus with soft tissue component infiltrating into it

and dyspnea. In this particular case, patient did not have any of these features except for occasional dull radiating pain in the shoulder. Brain, adrenal gland, liver, and bone are the common distant sites for a Pancoast tumor to be metastasized. To the best of our knowledge, there have been no previous reports of metastases of Pancoast tumors to the gingiva.

Though the adrenal gland is one of the common sites of metastasis of Pancoast tumor, it usually occurs unilaterally. However, bilateral adrenal metastases are seen in 10% of all lung cancer patients; of these 2–3% occurs at the time of initial presentation of non-small cell lung cancer.<sup>[5]</sup> Studies have shown that bilateral adrenal neoplasms are almost always metastatic tumors rather than primary, and clinical presentation varies with tumor type.<sup>[6]</sup> Hence, there was a need for search of a primary elsewhere in the body in our case, even after locating bilateral mass in the adrenal gland.

## CONCLUSION

To the best of our knowledge, as of now, there is no case report of simultaneous metastasis to both adrenal glands and oral cavity from a lung primary with the first symptoms noted clinically in the oral cavity and the patient being apparently healthy. As the prognosis of the metastatic lesions to the oral cavity is very poor, combination chemotherapy to alleviate the symptoms is the only preferred therapeutic modality. Because of its rarity, the diagnosis of metastatic lesions in the oral cavity is very often missed. Besides, it may simulate a benign process. So, diligent clinical and histopathological investigations should be done to diagnose the metastatic lesion and its origin.

## ACKNOWLEDGMENT

We would like to acknowledge the Department of Oral and Maxillofacial surgery, GDC, Trivandrum.

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**How to cite this article:** Beena VT, Panda S, Heera R, Rajeev R. Multiple metastatic tumors in the oral cavity. *J Oral Maxillofac Pathol* 2011;15:214-8.

**Source of Support:** Nil. **Conflict of Interest:** None declared.